Pulmonary Amyloidosis with Pulmonary Arteriovenous Fistula*

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A 77-year-old man was found to have multiple pulmonary amyloidoma and arteriovenous (AV) fistula of the lungs. Massive deposits of amyloid were present in the vascular wall, which apparently caused the fistula.

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Amyloid deposition has been reported to make vessels fragile, and some cases of hemorrhage caused by amyloidosis have been reported. We report herein a case of pulmonary amyloidosis suspected of causing a pulmonary AV fistula.

CASE REPORT

A 77-year-old man was admitted for close examination of abnormal shadows on his chest roentgenogram, which was taken during a routine check-up. His medical history revealed a gastric ulcer and cerebral hemorrhage. His family history was unremarkable. At the age of 62, his chest roentgenogram showed two tumorous shadows in the left upper lobe, and an open biopsy disclosed "benign lung tumors" of unknown origin. Since then he had received no treatment or follow-up examination. On admission, he was asymptomatic and his physical examination disclosed bruit on the right chest wall audible only on inspiration. No telangiectasia was seen on his skin or in his oral cavity. Laboratory data revealed increased ESR, CRP and CEA. Blood gas analysis showed mild hypoxia (PaO₂ of 62 mm Hg, PaCO₂ of 33 mm Hg) while the results of a lung function test were unremarkable. Sputum examination did not suggest malignancy or tuberculosis. Chest roentgenogram revealed bilateral multiple nodular calcified shadows (Fig 1).

Tomography revealed two vessel-like shadows connected to one nodular lesion in the right lower lobe, but no such shadows were found in the other nodular lesions. Pulmonary angiography showed an AV fistula in the right lower lobe, while the other nodules were unrelated to the pulmonary arteries (Fig 2). These nodules were thought to be benign because of their slow rate of growth. A right lower lobectomy was performed for resection of the AV fistula and biopsy of the other tumors. Pathologic examination showed the nodular lesions in the right lower lobe to be a solitary AV fistula with amyloid deposition. All the other nodules were amyloidomas. Massive amyloid deposits were seen in the vascular walls of the fistula (Fig 3), and the amyloid was found to be Λλ type. Further examinations (including rectal biopsy, gastroscopy, fiberoptic bronchoscopy, bone marrow aspiration) were performed, but no amyloid deposits were detected in other organs. No specific cause of amyloidosis was found. Re-examination of the old specimens disclosed that the first two tumors were also amyloidomas. The final diagnosis was primary amyloidosis of the lungs with a solitary AV fistula. After the operation, hypoxia disappeared and the patient has been well with no special treatment.

DISCUSSION

Amyloid deposition in the vascular wall is commonly seen in amyloidosis and is known to make vessels fragile.1

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Figure 1. Chest roentgenogram taken on admission; multiple nodular shadows, some of which were calcified, were evident.

Amyloidosis has been reported to cause massive hemorrhage in the skin, urinary bladder, gastrointestinal tract, and central nervous system.1,6 As for pulmonary amyloidosis, amyloid deposition in the vascular wall is well known in diffuse septal amyloidosis. Road et al7 reported a case of diffuse septal amyloidosis with massive hemoptysis caused by a ruptured pulmonary artery. Amyloid deposition in the vascular wall was regarded as having caused the rupture. In case of nodular amyloidosis, vascular deposition is also known,8 but few bleeding cases have been reported.

The relation between vascular deposition of amyloid and AV fistula is not well known. Walley9 reported a case of gastric AV fistula with amyloid deposition, but no definitive explanation about the relation between amyloid deposition and AV fistula was given.

Figure 2. Pulmonary angiography showing an AV fistula in the right lower lobe. Note that no other nodules were related to the pulmonary arteries.
Calcified Plaque in the Superior Portion of the Major Fissure*

An Unusual Manifestation of Asbestos Exposure

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Parietal pleural plaque is a well-recognized sign of exposure to asbestos. Visceral pleural involvement is an uncommon manifestation, and calcified visceral pleural plaques are rare. Those reported have been in the minor fissure and inferior major fissure. We describe a unique calcified plaque in the superior major fissure.

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Pleural plaque is a well-recognized sign of exposure to asbestos, occurring most often on the parietal surface. Visceral pleural involvement, recognized by interlobar fissure thickening, is an uncommon manifestation, although one group‡ reports that the incidence may be higher than is generally believed. The development of both parietal and visceral plaque is related to the duration of exposure to asbestos dust. Calcified visceral pleural plaques are rare. Those reported have been in the minor fissure† and inferior portion of the major fissure. To the best of our knowledge, this case report is unique in describing a calcified plaque in the superior portion of the major fissure.

CASE REPORT

This 63-year-old man presented to the hospital with symptoms of severe back pain; subsequent investigations revealed vertebral involvement by malignant histiocytic lymphoma. He had had exposure to asbestos during a 40-year career as a railroad worker and had a past history of chronic nonproductive cough and dyspnea on exertion. Putoroanterior and lateral chest roentgenograms taken on admission demonstrated hyperinflated lungs with perihilar thickening and plaque along the right lateral chest wall and hemidiaphragms.

A dense ovoid mass was present in the midportion of the right hemithorax. On the lateral view, this density projected along the

REFERENCES


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Calcified Plaque in Superior Portion of Major Fissure (Rupp, Jolles)